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# A REVIEW OF 3 PUZZLING CASES: OSTEOBLASTOMA-LIKE OSTEOSARCOMA

**Introduction:** The osteoblastoma-like variant is a very rare form of osteosarcoma. The differential diagnosis is critical since the treatment and prognosis are completely different for osteoblastoma and osteosarcoma. Clinical, radiological and pathological pitfalls are common. Three cases of osteoblastic osteosarcoma, who presented independently from each other with relatively short intervals and who were managed by the same multidisciplinary team, are retrospectively reviewed in this paper in regard to diagnostic difficulties and management.

**Patients and Method:** The first case was a 16 year-old male patient, who presented in February 2013 with pain and swelling in his right wrist. The radiological examinations revealed a lytic permeative bone lesion with contrast enhancement and soft tissue component in the distal radius. The lesion was also active on Tc-99 bone scan. Tru-cut biopsy was performed. Although there was no sign of malignancy in regard to cytology, focal areas of lace-like osteoid were observed. The case was consulted with a prominent international instituion and a mutual diagnosis of osteoblastoma-like osteosarcoma was reached. The patient underwent preoperative chemotherapy. Intraarticular wide resection of distal radius and biological reconstruction with vascular fibula including the fibular head were performed. The necrosis rate was 100% in the resected specimen and the surgical margins were negative. The patient completed adjuvant chemotherapy regimen. He was followed up for 27 months.

The second case was a 5 ½ years old female patient, who presented with left wrist pain in March 2013. The radiology revealed a lytic expansile lesion in the distal metaphysis of radius. Central sclerosis and heterogenous contrast enhancement were noted on MRI. Trucut biopsy yielded an active osteoblastic lesion with immature osteoid formation. A low-grade osteosarcoma was suspected and an open biopsy was performed to confirm

the first biopsy. However, histological features, which were typical of osteoblastoma, were noted. Unfortunately, rapid progression of symptoms necessitated a third biopsy. Evaluation of the third tissue specimen revealed a neoplasm that varied in morphology. including randomly organized immature osteoid. Osteoblastoma-like osteosarcoma was the definitive diagnosis and she underwent neoadjuvant chemoteraphy. Intercalary wide resection of distal radius and transposition of distal ulna to the radius were performed. The necrosis rate was 95% and the surgical margins were negative. Adjuvant chemotherapy was completed. She was followed up for 26 months.

The third case was a 27 year-old cardiology resident, who presented with constant pain in his right shoulder in July 2013. Radiology revealed a lytic-sclerotic cortical lesion at tuberculum minus. CT images were consistent with nidus formation and MRI demonstrated bone marrow edema surrounding central sclerosis. Focally increased uptake in the proximal humerus was evident in scintigraphy. The lesion was curetted with the radiological and clinical diagnosis of osteoid osteoma. The postop pathology was reported as benign osteoblastic lesion. The symptoms resolved as well. However, local recurrence with sclerotic appearance in radiographs was observed within 2 months. Excisional biopsy of recurring lesion yielded osteoblastoma. The rapid recurrence despite benign histology was found to be suspicious. Whole body bone scan was performed and a new lesion was noted in the left scapula. Scapular lesion, which was presumed as the metastasis of low-grade OS at this point, was resected immediately with wide margins. Taking into consideration the clinical progress and the histological findings, the diagnosis was revised as osteoblastoma-like osteosarcoma. This case was also consulted with a prominent international institution and the diagnosis was mutually agreed upon. Neoadjuvant chemotherapy was given. Then, he underwent wide resection of proximal humerus including closed glenohumeral joint resection. Tumor prosthesis reconstruction was performed. The necrosis rate was 90% and the surgical margins were clean. He also received adjuvant chemotherapy. He was followed up for 23 months.

**Results:** Full union was achieved in the first two patients, who underwent biological reconstruction. The implants were removed after union. The first two cases had no evidence of disease at the time of last follow-up while local recurrence was detected in the right shoulder of the third patient during the last follow-up visit. He recently underwent wide resection of the recurring mass. Second-line chemotherapy is being considered according to pathology result.

**Discussion:** The rarely occurring grey-zone between osteosarcoma and osteoblastoma creates a serious problem for diagnosis and consequently for determining the best therapeutic approach. Inconsistencies between clinical, radiological and pathological findings and unexpected behavior of the lesion must be carefully analyzed. High index of suspicion and experience are prerequisites for diagnosis.